disease are limited. However, Werner[9] recorded a patient with surgically inoperable Riedel's struma who had decided regression in the size of the gland after administration of full replacement doses of desiccated thyroid. In addition, some of the patients reported by Woolner and coworkers[10] had apparent cessation of the disease process while on thyroid therapy, although the data do not indicate whether or not full replacement doses were given. Conversely, that report also included several cases in which spontaneous regression followed limited and incomplete surgical excision. Indeed, the possibility of spontaneous regression of this disorder was stressed by Riedel himself.[6] However, the prompt diminution in thyroid size, noted in the present case after therapy, with desiccated thyroid, strongly suggests that the hormone was causative.

Recent studies indicate that enhanced endogenous secretion of thyrotropin is the cause of the goiter of Hashimoto's thyroiditis, and that perhaps such a mechanism is operative in other diffuse goiters. This evidence offers a rationale for the effectiveness of thyroid therapy in suppressing thyroid growth, since thyroid-hormone supplementation presumably would inhibit thyrotropin secretion and permit regression of the goiter. No such mechanism can be conjectured for Riedel's thyroiditis, however, since there are no data to support the concept of hyperthyrotropism in this disorder. The pathogenesis remains obscure and therefore any attempt to explain the therapeutic effect observed in this patient would be speculative.

Despite the inability to account for this effect physiologically, the observation is clinically important, if confirmed, since the results of the present method of treatment are not entirely satisfactory. If untreated, severe local-pressure symptoms often develop, and surgical resection is usually incomplete because of the extensive invasion of adjacent structures. Radical operation entails destruction of important contiguous tissue ensues. If these complications could be obviated by a trial of thyroid hormone with regression of the goiter, the benefit to the patient would be enhanced. However, this observation will need amplification through treatment of a large number of patients in this manner to determine if the response is universal. In addition, surgical biopsy of the thyroid gland will still be required to confirm the diagnosis.

Summary

A 47-year-old man with histologically proven Riedel's thyroiditis was observed to have a recurrence of the disorder nine months after operation. There was no evidence of hypothyroidism but after full replacement doses of thyroid hormone were given, the thyroid gland promptly returned to normal size. It is suggested that thyroid-suppressive therapy may be effective in Riedel's thyroiditis, as it is in Hashimoto's thyroiditis.

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Regional Enteritis in Siblings

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SINCE CROHN first described regional enteritis, a great deal of speculation has arisen as to the basic cause of the disease. Genetic factors have always been considered, but to date there has been no definite method to prove or disprove such factors. We have recently seen two siblings in whom the diagnosis of regional enteritis was made within a year of each other.

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Reports of Cases

CASE 1. The patient was a 15-year-old schoolboy who was admitted to hospital on January 10, 1963, with an eight-month history of low-grade diarrhea of three to four loose bowel movements a day, weight loss and weakness. In spite of a voracious appetite, the patient lost 15 pounds in weight. Just before admission he had begun to notice abdominal cramping usually relieved by a diarrheal movement with no blood or mucus in the stool. He had been having low-grade fever without night sweats.

The patient's parents had no history of gastrointestinal disturbances except that the mother had had colonic polyps removed. There was one other sibling, a brother, who is presented as Case 2. The patient had lived all his life in the San Francisco Bay Area.

Results of physical examination, including sigmoidoscopy, were within normal limits. For laboratory examinations see Table 1. Protein bound iodine was 4.8 micrograms per 100 ml.

No abnormalities were seen in x-ray films of the chest. A small bowel series showed multiple skip areas of abnormality involving the lower third of the ileum with abrupt change in the normal mucosal pattern and caliber to considerable narrowing and pronounced irregularity of the mucosal outline. There was extensive involvement of the distal foot of the ileum with persistent polypoid filling defects. These were thought to represent possible granulomatous polyp formation. (See Figure 1.)

Administration of belladonna, salicylazosulfapyridine (Azulfidine®) and high protein, low residue diet was prescribed. In a period of ten days the patient gained approximately five pounds and he was having only one loose, watery bowel movement a day.

The patient continued taking the prescribed drugs for four months. When seen approximately a year later, he was taking no medications but was following the diet and his weight had increased to over 125 pounds.

TABLE 1.—*Laboratory Examinations During Hospital Admission for Patients in Cases 1 and 2*

	Case 1	*Case 2*
Hemoglobin, gm per 100 ml	11.9	15.6
Hematocrit, per cent	36	49
Leukocytes per cu ml	4.900	6.800
VDRL test for venereal disease	Negative	Negative
Stools for ova and parasites	Negative	Negative
Carotene, micrograms per 100 ml	20	95
Albumin:Globulin ratio	3.8:2.7	4.4:2.7
Purified protein derivative	Negative	Negative
Urinalysis	Normal range	Normal range

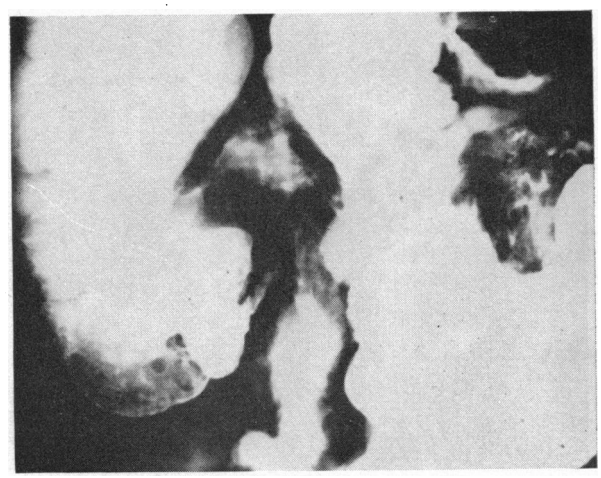

Figure 1.—(Case 1)—Barium enema study showing involvement of terminal ileum and skip areas proximally.

CASE 2. The patient was a 22-year-old electrician's helper who was admitted on January 6, 1964, because of recurrent episodes of alternating constipation and diarrhea (two to four loose, watery bowel movements a day) for two or three years. Diarrhea was accompanied by mild, lower abdominal cramping pain and then would gradually disappear. While serving in the armed forces in Vietnam the patient had had almost continuous diarrhea and had lost approximately 40 pounds in weight. During this time he also had malaria. He had been given various antidiarrheal medications by corpsmen but had never been seen by a physician. He said that he did not have night sweats. When seen before admission to hospital he complained of moderate weakness, easy fatigability and loose, watery bowel movements with occasional bright red blood in the stools.

Results of physical examination, including sigmoidoscopy, were within normal limits. A biopsy of the mucosa of the sigmoid colon showed no abnormalities. Results of laboratory examinations are shown in Table 1.

A roentgen series of the small bowel and a barium enema study showed "ragged" changes along with narrowing of the terminal ileum for a distance of about 7 to 8 centimeters proximal to the cecum. This was shown in both the barium enema and the small bowel series (Figure 2).

The patient was given a course of Azulfidine®, belladonna, multi-vitamin preparations, meprobamate as necessary for nervousness, and a bland, low residue diet. When seen in follow-up over the next two months he was asymptomatic, had discontinued all medication and at the end of that time had gained 5 pounds in weight.

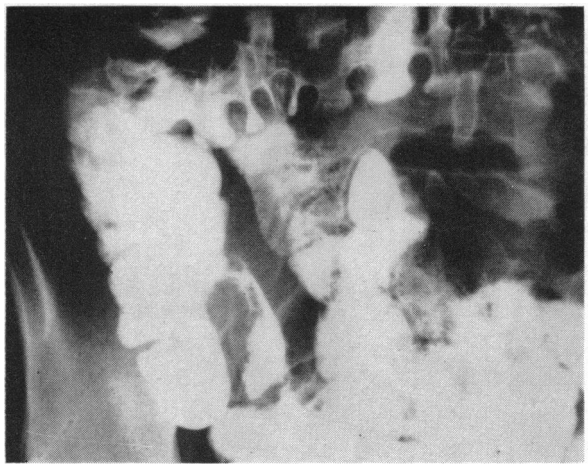

Figure 2.—(Case 2)—Barium enema study showing changes in terminal ileum consistent with regional enteritis.

Discussion

Sherlock and coworkers[3] reviewed the literature of the familial incidence of regional enteritis and ulcerative colitis as part of a study of a family with two male and two female siblings and one cousin who had regional enteritis, and two cousins who had ulcerative colitis. They listed a reported total of 30 families in which siblings had regional enteritis. In these families there were six sets of twins, four of which were monozygotic and two were dizygotic. The prevalence of ulcerative and regional enteritis in hospital studies of all patients admitted was 0.3 per cent. The chance probability of two siblings having the disease, based on this 0.3 per cent, was 1 to 111,000 or 0.000009 per cent. The probability that three members of the same family would be affected would be 1 in 37,000,000. Hence Sherlock felt that the genetic factors were more important than environmental factors, but that no conclusion was possible as to the genetic mechanism because of the very small number of cases studied to date.

Crohn and Yarnis[1] reported a total familial incidence of 5 per cent in their large series of cases of regional enteritis. Felson and Wolarsky[2] found 21 family groupings with a total of 38 cases. In their study, 3.1 per cent of 1,204 patients with regional enteritis or ulcerative colitis were in sibling groupings.

In studying the various family groups in which the disease has been shown to occur, little evidence was found of a simple, single dominant or recessive gene as the most likely method of transmission of the disease.

Summary

Two cases of regional enteritis occurring in siblings within one year of each other have been presented. Statistical data in other reports in the literature indicates a genetic factor is involved in the etiology.

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